Trigeminal sensory neuropathy and rheumatoid arthritis: case study of a rare association

Sir, Isolated trigeminal sensory neuropathy (TSN) is a distinct entity of heterogeneous aetiology. A chronic form of TSN has been associated with connective tissue disorders (CTD) [1–7], but TSN has rarely been reported to develop as an isolated neurological deficit in patients with rheumatoid arthritis (RA) [2, 4]. We present one case of TSN in a patient with RA.

A 40-yr-old lady was first seen in our hospital in 1979 with an inflammatory polyarthritis of RA type. Tests for rheumatoid factor were negative but the clinical course was typical of RA, with symmetrical polyarthritis and the development of progressive joint erosions. In March 1991 the patient developed numbness affecting the left lower lip. Over the next few weeks, numbness extended over the whole of the left side of her face and then affected the right side of the face up to the maxilla. Initially she would inadvertently bite her tongue and cheek. There was no drooling, difficulty in speaking, psychiatric disturbances, interference with sexual function, sphincter disturbances or dryness of mucosa. On sensory testing (light touch, pinprick pressure and
thermal discrimination) she was found to have complete bilateral sensory impairment in the maxillary and mandibular divisions of the trigeminal nerves. Corneal reflexes were normal, the muscles of mastication were intact and there were no other cranial or peripheral nerve abnormalities. Examination of the head, neck and oral cavity was normal. Investigations showed positive tests for rheumatoid factor (Rose-Waaler test, 1:16), weakly positive IgG antibodies to nuclear factor (1:80) and negative anti-DNA antibodies. Fasting blood glucose, calcium and liver function tests were within normal limits.

X-rays of the cervical spine showed a minor degree of atlanto-axial subluxation at 4 mm but no evidence of vertebral subluxation; a CT scan of her brain demonstrated normal findings. Her symptoms and signs of TSN had remained unchanged over 9 yr. Her arthritis had progressed slowly, with predominant involvement of the hands and feet, but she had not developed any other extra-articular manifestations of RA and her symptoms were reasonably controlled on combination therapy (sulphasalazine and hydroxychloroquine) and diclofenac.

This case shows features of chronic isolated TSN in association with RA. As with other cases of TSN associated with CTD, the presentation was subacute and bilateral, and there was involvement of more than one division of the trigeminal nerve [1–3, 5–7]. Tongue biting, as found in this case, has been reported commonly. The trigeminal motor pathway is usually spared [1], as seen here, and this is demonstrated by sparing of the muscles of mastication. Facial pain can be a prominent and distressing feature of this condition but lancinating facial pain should alert the physician to the possibility of trigeminal neuralgia. Although in TSN there may be involvement of other cranial nerves, this was not the case in our patient.

The pathogenesis of TSN is unknown, although it is considered to be either autoimmune, from its association with CTD and diabetes mellitus, or vasculitic, on the basis of post-mortem studies and peripheral nerve biopsies [8]. The site of the lesion is probably in the peripheral nerve, although the evidence to date is not conclusive.

The differential diagnosis of TSN is wide and in many cases remains unexplained [9].

Patient interview, physical examination and sensory testing are used to evaluate, document and monitor nerve dysfunction. Complete examination of the head, neck, postnasal space and oral cavity should be performed as well as the appropriate plain radiographs, CT or MRI scan of the brain. Fasting blood glucose, plasma calcium levels, autoimmune screen and liver function tests would complete the diagnostic investigations. A lengthy duration of symptoms over a period of several years with little change or improvement is often found [2, 8]. The severity, onset and duration of symptoms are independent of disease activity and TSN can precede a clinical diagnosis of a CTD by several years [2, 9].

A. Vilches, M. J. Burke

Burnley General Hospital, Casterton Avenue, Burnley, Lancashire BB10 2PQ, UK

Accepted 19 January 2001

Correspondence to: A. Vilches-Moraga, 38 Healdwood Drive, Burnley, Lancashire BB12 0EA, UK.